Head and Neck Pathol (2009) 3:283–285
DOI 10.1007/s12105-009-0141-2

Clinico-Pathologic Conference: Case 1

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Received: 4 September 2009 / Accepted: 28 September 2009 / Published online: 14 October 2009

Clinical Presentation

A 42-year-old Caucasian female presented with extreme pain of the right mandible and paresthesia of her right lower lip from the commissure to the midline. The patient reported that sudden severe pain began 4 months earlier in the right mandible. Consultation with a private dentist for the pain resulted in the initiation of endodontic therapy for tooth #30. Subsequent to the completion of root canal treatment on tooth #30, the pain increased and the paresthesia began. Both the pain and paresthesia were non-responsive to a short course of steroids and antibiotics. Tooth #30 was extracted 1 month prior to presentation. Following the extraction, the pain and paresthesia worsened and a "swishing" sound developed in her right ear. Evaluation by otolaryngology was negative. Intraoral and extraoral examinations were unremarkable except for the right lower lip paresthesia. Panoramic (Fig. 1), occlusal (Fig. 2) and periapical radiographs (Fig. 3) were taken at the time of presentation.

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Differential Diagnosis

The panoramic radiograph revealed a poorly defined radiolucent area extending from the right mandibular second premolar area to the right mandibular third molar. There was evidence of widening of the mandibular canal (Fig. 1). The cross sectional occlusal radiograph revealed a radiolucent area predominantly in the mandibular right first and second molar region. There was no evidence of cortical expansion or periosteal reaction (Fig. 2). A periapical radiograph revealed a poorly defined radiolucent area extending from the right mandibular second premolar to the right mandibular third molar area. Evidence of the recent extraction of #30 was present. There was no evidence of root resorption, but the lamina dura was only partially intact on both the second premolar and the second molar (Fig. 3). An axial computed tomographic image revealed an osteolytic lesion distal to the right mandibular second premolar. There was fenestration of the lingual cortex without evidence of expansion of either cortex. The CT scan was interpreted by her physicians as possibly representing "abscess formation" (Fig. 4). Although an acute infection of pulpal origin can produce poorly defined radiolucencies of bone and symptoms of pain and paresthesia, [1] an infectious etiology was ruled out when symptoms failed to resolve after extraction, antibiotics and corticosteroid therapy. Inflammation and infection related paresthesia is thought to be produced by mechanical pressure and ischemia associated with the inflammatory process [2].

Based on the clinical history and radiographic findings at the time of presentation, a differential diagnosis was formulated that included metastatic carcinoma, lymphoma, peripheral nerve sheath tumor and vascular malformation. All of the clinical and radiographic findings, including the

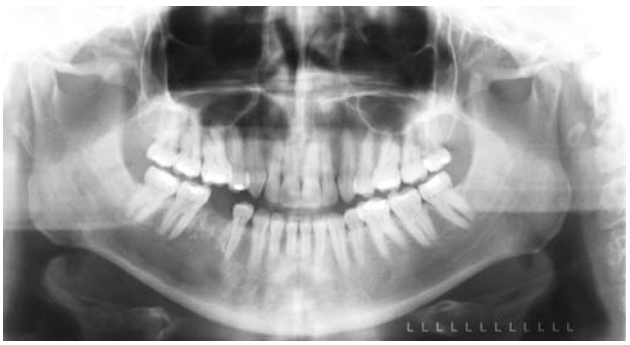

Fig. 1 Panoramic radiograph

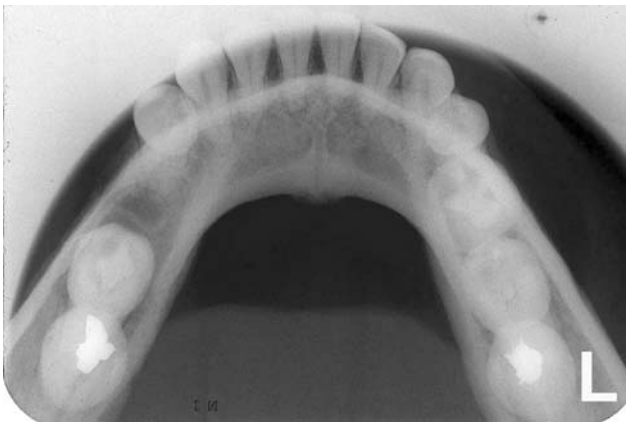

Fig. 2 Occlusal radiograph

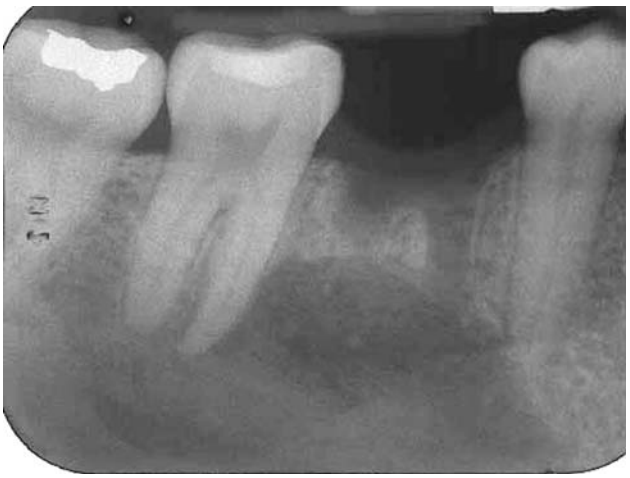

Fig. 3 Periapical radiograph

widening of the mandibular canal, can be encountered in these pathologic states. Metastatic carcinoma to the mandible produces varied clinical and radiographic signs and symptoms. Deposits of metastatic carcinoma most often produce ill-defined radiolucencies of the mandible. External root resorption can be seen with metastatic disease, but

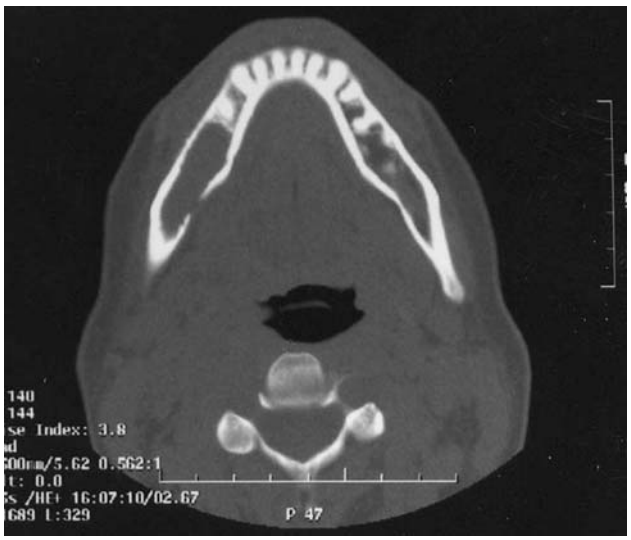

Fig. 4 Axial CT

was not present in this case. Pain, swelling, loosening of teeth and paresthesia are the most common presenting symptoms. Paresthesia of the lower lip and chin (numb chin-syndrome) may also be encountered [1]. Extranodal non-Hodgkin's lymphoma (NHL) of the jaws is most often a component of widely disseminated disease, although there are reports of primary mandibular involvement of isolated NHL [3]. Lesions of the mandible may produce complaints of pain and paresthesia, including numb chin-syndrome. There are no pathognomonic radiographic findings, but most often intraosseous NHL results in an ill defined radiolucency, which can be associated with loss of lamina dura and widening of the mandibular canal [4]. Benign neural or vascular tumors originating from the neurovascular bundle within the mandibular canal may widen the canal and cause paresthesia, which are findings more commonly associated with malignant processes. Intraosseous neurolemoma (schwannoma) of the mandible has a varied radiographic and clinical presentation, but most often presents as a well defined radiolucency. Widening of the mandibular canal and cortical thinning have been reported, as has the frequent direct association with the neurovascular bundle seen intraoperatively [5]. Paresthesia and pain are not unusual for intraosseous schwannomas [1]. The radiographic presentation of intrabony vascular malformations is highly variable. Most often a multilocular radiolucency is seen, but the radiographic variation is wide with reports of arteriovenous malformations (AVM) producing ill defined radiolucencies, loss of lamina dura, cortical thinning, and widening of the mandibular canal. Symptoms of pain and paresthesia of the lower lip have been reported with AVM of the mandible [6].

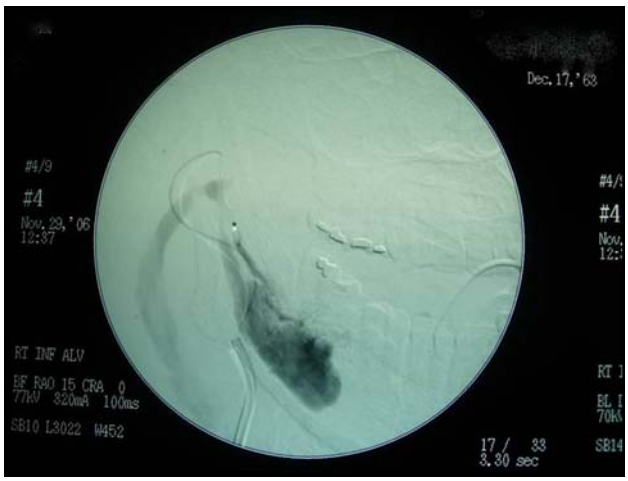

Fig. 5 Digital subtraction angiogram

Diagnosis and Discussion

The patient underwent surgical exploration in the operating room at the Hospital of the University of Pennsylvania. There was no gross hemorrhage on aspiration. A 2 mm window in the buccal cortex revealed a high flow vascular malformation. After hemostasis was achieved, the patient was transported from the operating room to interventional radiology where an emergency angiogram was performed. The digital subtraction angiogram revealed an abnormal vascular structure in the inferior alveolar canal region which was consistent with a high flow vascular malformation largely supplied by the inferior alveolar artery (Fig. 5). Interventional radiology was able to achieve almost complete occlusion of the lesion with "glue". The patient reports that her paresthesia resolved completely within 6 months of the occlusion procedure. The patient has fully recovered with no evidence of residual or recurrent vascular malformation.

Vascular malformations occur as a result of late errors in embryogenesis and although they are present at birth, they typically go undiagnosed for many years [7]. High flow vascular malformations are associated with significant morbidity and mortality secondary to uncontrollable bleeding [8]. Although rare, 50% of intrabony vascular malformations occur in the maxillofacial region, where they most commonly become clinically apparent in the second decade of life [9]. The presenting symptom in the jaws is most often spontaneous gingival bleeding [10]. Other less common and less characteristic symptoms include pain, swelling and earache. There have been several reports of paresthesia associated with mandibular arteriovenous malformations [6]. Although the radiographic presentation is not pathognomonic, vascular malformations of the jaws most often present as multilocular radiolucencies on plain films [1]. The present case is an example of a high flow vascular malformation of the mandible presenting as a poorly defined radiolucency in a woman two decades older than the average reported age at presentation with the uncommon chief complaint of pain and paresthesia. This case supports the inclusion of vascular malformations in the differential diagnosis of ill defined radiolucencies of the jaws; their consideration allows for proper surgical planning that minimizes potentially life threatening complications.

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